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FIBROUS DYSPLASIA OF THE JAWS: A REVIEW OF LITERATURE
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FIBROUS DYSPLASIA OF THE JAWS: A REVIEW OF LITERATURE

D.O. AWANGE

SUMMARY

This communication reviews the literature on fibrous dysplasia of the jaws particularly the available published reports on this condition from Africa. It reveals that few cases appear to have been reported from Africa, reflecting the lack of research work and case reports. The majority of these cases are generally reported in advanced stages indicating the late presentation for treatment and delayed diagnosis. Information regarding follow-up records appears scanty and this probably may have contributed to the difficulty in determining the prognosis and any recurrent rate. Much effort still needs to be directed at research on oral lesions to determine their true occurrence and distribution in the African continent.

INTRODUCTION

The term "fibrous dysplasia" was introduced by Lichtenstein in 1938 to designate a group of single or multiple bone conditions which were previously put under many titles such as osteitis fibrosa, osteodystrophica fibrosa, osteitis ossea, fibrous osteoma, hypertrophic localised osteitis and localised leontiasis ossea(1,2). It is defined as a benign bone lesion characterised by the replacement of the interior of the normal bone by the abnormal proliferating connective tissue exhibiting varying amount of osseous metaplasia(3,4,5). Fibrous dysplasia of bone may be monostotic, when a single bone is involved; or polyostotic when several bones are involved. Occasionally, it may occur as part of a complex known as Albright's syndrome, with symptoms of polyostotic lesions, abnormal melanotic skin pigmentations or cafe au lait spots, sexual precocity, premature skelcton maturation and occasional hyperthyroidism(3,4,6). When polyostotic lesions are limited only to the bones of the craniofacial complex, the condition is referred to as craniofacial fibrous dysplasia(7). Clinically, fibrous dysplasia may exhibit a slow-growing and painless swelling of the affected part producing deformity; the growth often slows or ceases after the onset of puberty(4,5). In certain conditions such as pregnancy, it may be reactivated(5,6). It is non-encapsulated and essentially benign both in its histological components, and clinical and biological course(8). Research work and literature reports on fibrous dysplasia of bone including the jaw bones have been extensively carried out in other parts of the world especially the western world. Although remarks have been made by several authors to the effect that fibrous dysplasia of the jaws is more common in West Africa than in the whole continent of Europe(9,10), its true nature of occurrence and distribution in the African continent still appears uncertain. The purpose of this paper therefore, is to analyse and discuss the occurrences, distribution and clinical features of fibrous dysplasia of the jaws from Africa by reviewing the literature reports including the available published reports on this condition from Africa.

RESULTS

The available published literature reports on fibrous dysplasia of the jaws from Africa were obtained form the Royal Hallamshire Medical and Dental Library, University of Sheffield, UK in 1987 and the British Council Library, Nairobi, Kenya, 1990.

The clinical findings from these reports are summarised (Table 1). The table covers information about the source, year of publication, country of origin, age, sex and site distributions. Other findings not tabled but which are also summarised include signs and symptoms, Albright's syndrome, malignancy in fibrous dysplasia, radiological and histological features, treatment, follow-up and recurrence or reactivation. The cases listed (Table 1) have therefore been accepted on the basis of information including documentations such as radiographs, photomicrographs and clinical descriptions that appear to have sufficient evidence diagnostic of fibrous dysplasia of bone(4,5). The majority of these reports (Table 1) appear to have come from West Africa particularly Nigeria. They are mainly from English-speaking African countries and hence more readily available. Reports from non-English speaking African countries (French, Arabic, Portuguese) are not included because of the unavailability of specific information on studies carried out in this condition in those countries.

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Age, sex and site distributions: The age distribution shows that the mean ages of the patients from the reported cases of fibrous dysplasia of the jaws from Africa ranges from 16.5-33.0,with a peak age mostly in the second and third decades of life; the youngest patient is four years and the oldest sixty nine years (Table 1). The sex distribution varies from a higher prevalence in females than males, showing a ratio of 3:1, to an equal sex distribution, or even in a few instances of more cases in males than females. The site distribution generally shows more cases in the maxilla than the mandible.

Signs and symptoms: The signs and symptoms in the reported cases of fibrous dysplasia including all forms — monostotic, polyostotic and craniofacial reported from Africa are generally the results of late presentations for medical attention. They consist of progressive large swellings of the jaws, face or skull with disfiguring sizes and disorganization of normal architecture of the affected regions. Pain is not a feature unless a vital organ is interfered with. Teeth are displaced, functional impairments include speech, feeding, nasal obstruction, proptosis, diplopia, headaches, strabismus with disseminated visual acquity and in a few cases deafness and blindness.

Albright's syndrome: From this review, there was only one case of Albright's syndrome. It was among the forty seven cases of fibrous dysplasia which also included forty three monostotic cases, and three polyostotic cases(9). The case involved a 28 year-old male with a deformity of maxilla, skull, limb bones and a melanotic pigmented patch on the anterior abdominal wall.

Malignancy in fibrous dysplasia: Among the reported African cases, only two are reportedly associated with malignancy. One case involved a Sudanese aged 25 years who had fibrous dysplasia of the maxilla with squamous cell carcinoma thought to have arisen form

Table 1

Reported cases of fibrous dysplasia of the jaws from Africa

Source, Year, Country	Cases	Age			Sex		Site		
		Range	Mean	Peak	Male	Female	Mandible	Maxilla +	Cranio-facial
Odeku et al (8) (1969) Nigeria	27	5-42	18.0	-	16	11	3	15	9
Daramola *et al* (9) (1976) Nigeria	47	5-65	25.0	11.20	12	37	18	29	-
Williams and Faccini (11) (1973) Nigeria	5	7-43	21.0	-	2	3	-	5	-
Williams *et al* (12) (1974) Nigeria	13	8-38	21.3	2nd decades	3	10	7	6	-
Adekeye *et al* (13) (1980) Nigeria	13	6-29	16.5	-	7	6	7	6	-
Ajagbe *et al* (14) (1982) Nigeria	66	-	-	-	26	40	14	45	7
Ajagbe *et al* (15) (1983) Nigeria	98	4-68	27.17	11.20	42	56	35	56	7
Subbuswamy and Shamia (16) (1981) Nigeria	21	5-50	28.1	-	18	11	-	-	-
Obisesan *et al* (17) (1977) Nigeria	25	6-29	-	11.20	8	17	-	-	-
Anand *et al* (18) (1967) Nigeria	4	-	22.0	-	3	1	1	3	-
Chatre *et al* (19) (1953) Ghana	1	-	21	-	-	1	-	-	1
Samy *et al* (1) (1967) Egypt	7	14-26	28.7	-	6	1	-	7	-
Sidney (20) (1951) S. Africa	1	-	21	-	-	1	-	1	-
Lellow and Sparrow (7) (1985) S. Africa	3	15-22	17.7	-	2	1	-	-	3
Viljoen et al (21) (1988) S. Africa	1	-	-	-	1	-	-	-	1
Chindia (22) (1990) Kenya	1	-	33.0	-	-	1	1	-	-

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the antral mucosa(1). The other case is described to have become malignant after multiple surgical treatment(9).

Radiological features: The radiological appearances of fibrous dysplasia in the reported African cases are varied and include a cyst-like radiolucency, some being unilocular or multilocular with a "soap-bubble" appearance; and lytic and sclerotic areas usually with indistinct borders.

Obisesan *et al*(17), in an analysis of radiographs of 25 patients in Nigeria with histologically proven craniofacial fibrous dysplasia, classified the lesions into six radiological types, namely: orange-peel type; whorled plaque-like; diffuse sclerotic; cyst-like multicular or unilocular; pagetoid and chalky types. The orange-peel type was by far the most common (40%) followed by the plaque-like types (20%), cyst-like (16%) and sclerotic types (12%).

Histological features: The histological features in the reported African cases are typical of fibrous dysplasia as described elsewhere(4,5,8). Both in mature and young forms showed either woven bone or woven bone with occasional lamellar trabeculae undergoing active osteogenesis and lying in an abundant fibrocellular stroma; mature and sclerosed forms showed dense bony trabeculae or lamellar trabeculae with osteoblastic rimmings, lying in a sparse stroma. The histological appearances depended on the age of the lesions.

Treatment, follow-up and recurrence or reactivation: The basic principle for treatment for the African cases was surgical, mainly for cosmetic restoration, pain relief and relief of normal functional interferences such as nasal or oral obstructions. In advanced cases which were common, radical or aggressive excisions were performed while on smaller cases, curretages were done.

Follow-up records from the African reports were rather scanty or sketchy. In the series of Ajagbe *et al*(15), 13 patients were followed up to over 5 years; multiple operations due to recurrences were performed on 11 patients, one of whom was a 35 year old woman who had radical hemi-maxillectomy three times and whose lesion recurred during two successive pregnancies after the initial excision. In the series of Odeku *et al*(8), the periods of the follow-up ranged from a few months to six years. Recurrence was noted in a 5 year old boy, two years after excision in the right maxilla. Chindia(22) recently reported a case in a 33 year old female presenting with an apparently reactivated fibrous dysplastic jaw lesion, with evidence of cystic degeneration. He noted that while conservative surgical procedures remain the treatment of choice, situations may arise where tremendous tissue activity would require early intervention by numerous cosmetic surgical shaving procedures. Continued follow-up for this patient was not possible to determine the full prognosis of the disease.

DISCUSSION

Fibrous dysplasia is a congenital, metabolic, non-familial disturbance that produces 2.5% of all tumours and over 70% of all non-malignant tumours of the bone(23). This literature review, based on the available published literature reports on fibrous dysplasia of the jaws from Africa, reveals that quite a few number of cases appear to have been reported from Africa, most of which are from West Africa, particularly Nigeria. Although the results of this review tend to reflect the general lack of enough research work on this condition in African continent, its slight predominance in Nigeria over other parts of Africa may probably be attributed to first, more research on it may have been carried out in Nigeria and reported in the literature that in other parts of Africa; secondly, it may also be probably due to other contributing factors such as environmental, hereditary and dietary factors. This, therefore, is an area which may require further investigation, since several authors(9,10) have even remarked that fibrous dysplasia are more common in West Africa than in the continent of Europe. As regards age, sex and site distribution, the results of this review are in agreement with others elsewhere(24,25,26). The majority of the patients are young, mostly in their second and third decades of life; more cases are in the maxilla than mandible. Females are generally more affected than males except in a few instances where both sexes were equally affected or more are in males than females.

Only one case of Albright's syndrome with skeleton abnormalities and cutaneous involvement was described in the review(9). It is estimated that for every case of Albright's syndrome there are thirty to forty cases of fibrous dysplasia manifested simply by bone involvement(6,27,28). Malignancy in fibrous dysplasia is very rare; it is benign and non-neoplastic lesion.

From this review, only two cases are reportedly associated with malignancy(1,9). Lichtenstein and Jaffe(3) pointed out in their study of the disease that they knew of no cases of fibrous dysplasia of bone in which any of the lesions had undergone malignant transformation, irrespective of skeleton involvement. Ekper(30) raised the possibility of sarcomatous transformation in fibrous dysplasia as a result of undergoing multiple operations and irradiation therapy. He was commenting on a case described by Ramsay *et al*(31) that had presented with a progressive fibrous dysplasia of the maxilla in a 3 year old boy with grotesque deformity, and had died at the age of 16 years after unsuccessful attempt at surgical resections.

The varied radiological appearances described in this review depend very much on the maturity of the lesions and the proportions of mineralised tissues present. Lesions that are chiefly fibrous or fibroblastic appear radiolucent or less radiopaque than those in which calcified areas and bone trebaculae predominate;

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a small proportion of these mixed lesions with dense bone appear sclerotic(23).

Treatment of fibrous dysplasia appears to depend upon several factors that include the growth rate, degree of deformity and dysfunction, age and health of the patient and expectations of the patient and the surgeon(33). For these asymptomatic patients in whom the lesions are slow-growing or have stopped growing, expectant therapy is preferred. For those symptomatic patients in whom the lesion is aggressive with tremendous tissue activities resulting in serious deformity, surgery must be considered. The obvious indications for surgical therapy therefore involve corrections of deformity, treatment of pathological fractures, relief of pain, or arrest of excessive growths. Surgery may also reduce the dangers of vision, hearing and speech caused by the encroachment of these tumours(23,33). But for aesthetic reasons, some form of reconstructive procedures with the use of bone grafts or implants should be performed after radical resections of aggressive deforming growths. In children, the feature of normal mandibular growth without deformity should not be overlooked in the preoperative surgical planning(34). Treatment with radiotherapy is, however, contraindicated as this can possibly damage the growth centres and furthermore, may lead to malignant degeneration(23,35,36).

A few cases were described as recurring or reactivated, some several times after treatment, even after the skeletal bone growth should have ended. This however appears unusual, and since the factors that cause fibrous dysplasia are unknown, and since bone is not static, this unusual growth may not be too surprising for conditions such as pregnancy has been well known to stimulate or reactivate the growth of fibrous dysplastic lesion(5,6,9,15,23).

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